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Early Language Development in Infants and Toddlers with Fragile X Syndrome: Change over Time and the Role of Attention

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Abstract

Fragile X syndrome (FXS) is associated with significant language and communication delays, as well as problems with attention. This study investigated early language abilities in infants and toddlers with FXS ($n = 13$) and considered visual attention as a predictor of those skills. We found that language abilities increased over the study period of 9 to 24 months with moderate correlations among language assessments. In comparison to typically developing infants ($n = 11$), language skills were delayed beyond chronological age- and developmental level-expectations. Aspects of early visual attention predicted later language ability. Atypical visual attention is an important aspect of the FXS phenotype with implications for early language development, particularly in the domain of vocabulary.

Keywords

visual attention; language development; fragile X syndrome

Attention deficits are highly prevalent in the fragile X syndrome (FXS) phenotype (Sullivan et al., 2006), and they are perhaps “the most striking and consistent primary behavioral features” of young children with FXS (Cornish, Sudhalter, & Turk, 2004, p. 12). As such, difficulties with attention may be a critical constraint on development in individuals with FXS (Cornish, Cole, Longhi, Karmiloff-Smith, & Scerif, 2012). As the leading inherited cause of intellectual disability, FXS results in significant language impairments; yet, very little research has focused on early development in infants and toddlers with FXS as language emerges (Abbeduto, Brady, & Kover, 2007; Bailey, Hazlett, Roberts, & Wheeler, 2011) or on specific predictors of later language development (Luyster, Seery, Talbott, & Tager-Flusberg, 2011). The goal of the current study was to contribute to what is known about attention and language in individuals with FXS by examining visual attention as a predictor of language and communication abilities over time in infants and toddlers with FXS. Identifying correlates and predictors of language and communication development in

very young children with FXS will be fundamental to informing theoretical perspectives on the cognitive processes that underlie atypical trajectories of language acquisition.

Early Language in Individuals with FXS

Relative to other neurodevelopmental disorders, such as autism spectrum disorder (ASD), for which a heavy emphasis on early diagnosis has been placed (Landa & Garrett-Mayer, 2006; Mitchell et al., 2006; Ozonoff et al., 2010), most research on language in individuals with FXS has focused on older children or included children from a wide range of ages. For example, a series of studies has characterized language abilities during the preschool and young school-age years (Roberts et al., 2001; Roberts, Mirrett, Anderson, Burchinal, & Neebe, 2002). Roberts et al. (2001) found that boys with FXS were characterized by large individual differences in language skills, with slow increases in abilities over the study period and even slower growth for expressive than receptive language. In a follow-up study, Roberts et al. (2002) examined aspects of communication development beyond receptive and expressive language in 22 males with FXS (21 to 77 months of age). In this smaller sample of boys with FXS, a profile of stronger verbal and vocal communication (e.g., sound and word use) relative to gesturing, reciprocity, and symbolic behaviors was identified, despite overall delays. This research has provided evidence of delayed language ability and wide variability among children with FXS, but because these studies included children across very large age ranges (e.g., 20 – 86 months), the extent to which conclusions about specific aspects or periods of early development can be made is somewhat limited.

Although comprehensive studies of early language development in individuals with FXS are lacking, some studies have provided estimates of timing for language milestones and early delays. Overall, the existing research suggests that developmental delays, in general, and delays in language development, in particular, are apparent well before the third birthday of children with FXS (Bailey et al., 2011; Baranek et al., 2008). Based on survey data from 249 families with a child with FXS diagnosed between 2001 and 2007, Bailey, Raspa, Bishop, and Holiday (2009) reported that a developmental delay was confirmed for boys with FXS at approximately 20 months on average. Regarding language delays, Mirrett and colleagues found that 6 of 11 boys with FXS were delayed on a language screening measure at 9 months, with 12 of 12 delayed by 12 months of age (Mirrett, Bailey, Roberts, & Hatton, 2004). Aligning with results from Roberts et al. (2002), a retrospective video analysis of seven infants with FXS between 9 and 12 months of age suggested that communicative functions and gestures may be limited in this population, with communicative attempts occurring primarily nonverbally (Marschik et al., 2014). There is converging evidence that the age of first words is substantially delayed in males with FXS, with an average age of 26 to 28 months (Hinton et al., 2013; Roberts, Mirrett, & Burchinal, 2001). In the context of these delays, the trajectories of development for specific aspects of language ability remain poorly understood.

Only a few studies have characterized the skills of infants and toddlers with FXS with an eye towards potential predictors of language ability. Considering gestures, Flenthrope and Brady (2010) found that increased gesture use by the child unexpectedly predicted lower vocabulary ability two years later in young children with FXS, whereas Hahn et al. (2014)

found that maternal gestures were positively associated with better later language in children with FXS. A series of studies focused on the responsivity of mothers of young children with FXS and demonstrated that early developmental level, autism symptoms, and maternal responsivity predicted vocabulary ability through age nine (Brady, Warren, Fleming, Keller, & Sterling, 2014; Warren et al., 2010). Of note is that Warren et al. found that these 55 children with FXS, who were between 11 and 48 months at the time of first assessment, significantly improved in receptive and expressive language abilities, but with some evidence of slowed growth over time. Roberts and colleagues (2009) examined change on a single language measure from 9 to 68 months for 55 boys with FXS (some participants were also included in the study by Mirrett et al. [2004]) and also found that autism symptoms were negatively associated with development. No study has considered specific aspects of cognitive function, such as attention, as a predictor of early language ability in individuals with FXS.

The current study extends findings from Mirrett et al. (2004) and Roberts et al. (2009) by characterizing both the extent of delay and observed change in language and communication ability across infancy and toddlerhood for boys with FXS using multiple assessments, including one measuring receptive and expressive vocabulary. The present work further builds on previous research by examining a specific aspect of development that is both known to be impaired in individuals with FXS and theoretically important to the development of language—visual attention—as a predictor of language and communication through 24 months of age.

Attention as a Predictor of Language Ability

With respect to both typical development and neurodevelopmental disorders, attentional processes likely play a critical role in the developmental trajectories of infants and children through multiple interacting mechanisms, including properties of stimuli in the environment, central nervous system maturation, and individual differences, such as genetic factors (Scerif, 2010). As such, visual attention may be pivotal in explaining how language and communication develop during infancy and through childhood in individuals with FXS. Evidence regarding impaired attentional processing in those with FXS and the relationship between attention and language in children with typical development or other neurodevelopmental disorders support this viewpoint.

Attention in individuals with FXS

The known cognitive profiles of children and adolescents with FXS include difficulties with particular aspects of cognitive processing, such as attention and executive function (Hooper et al., 2008; Lanfranchi, Cornoldi, Drigo, & Vianello, 2009; Ornstein et al., 2008). In fact, an estimated 84% of boys with FXS are diagnosed with or treated for attention problems (Bailey, Raspa, Olmsted, & Holiday, 2008). For boys with FXS, some attentional processes (e.g., gaze shifts between a person and object) may be a strength relative to other cognitive skills (Roberts et al., 2002), but visual attention in general is a weakness relative to typical development (Scerif, Longhi, Cole, Karmiloff-Smith, & Cornish, 2012). Aspects of visual attention predict later nonverbal cognitive abilities in boys with FXS and improve somewhat

over time during childhood (Cornish, Cole, et al., 2012; Scerif et al., 2012). However, switching attention between target types and inhibiting repetition may be weaknesses in children and adolescents with FXS (Wilding, Cornish, & Munir, 2002). Indeed, it has been suggested that early impairments in aspects of visual control could be a precursor or early developmental marker of the cognitive profiles observed later in childhood (Scerif et al., 2005).

Indicators of impaired visual attention in the FXS phenotype are also present in infancy and toddlerhood. For example, perseverating may be a signature of visual attention in two- to four-year-olds with FXS, reflecting poor inhibition (Scerif, Cornish, Wilding, Driver, & Karmiloff-Smith, 2004). Roberts, Hatton, Long, Anello, and Colombo (2012) found that infants with FXS displayed an atypical pattern of visual attention: 12-month-olds with FXS had increased durations of looking to a stimulus (i.e., a toy) relative to typically developing infants of the same age. In addition, Roberts et al. reported that infants with FXS demonstrated latencies to disengage their attention from the toy that were twice as long as the age-matched typically developing infants. Although not statistically significant, this pattern of disengagement may be similar to the difficulty with nonsocial orienting—particularly disengaging attention away from a stimulus—that is associated with ASD (Elison et al., 2013; Landry & Bryson, 2004; Renner, Klinger, & Klinger, 2005; Zwaigenbaum et al., 2005). Interestingly, attentional processing (i.e., increased duration of looking, increased latency to disengage) was associated with symptoms of ASD in this sample of infants with FXS. This study established that aspects of visual attention in infants with FXS are distinguished from typical development early in life, with potential consequences for later development.

Attention and language in typical and atypical development

Studies of typically developing infants and children with neurodevelopmental disorders, such as ASD or Down syndrome, suggest that aspects of attention could have important implications for development in multiple cognitive domains, including language (Adamson, Bakeman, Deckner, & Ronski, 2009; Cornish, Steele, Monteiro, Karmiloff-Smith, & Scerif, 2012; Karrass, Braungart-Rieker, Mullins, & Lefever, 2002). This research has hailed from both from an information-processing perspective, relying on infant habituation or other direct assessments using experimental measures, and a temperament perspective, relying on parent report (Colombo et al., 2009; Kannass & Oakes, 2008). Regardless of the approach, the majority of research has linked aspects of attention to vocabulary development in particular. Vocabulary acquisition begins extremely early in life, with typically developing infants demonstrating noun comprehension as young as six months of age (Bergelson & Swingley, 2012), meaning that early attentional processes have the potential to directly impact nascent development in this language domain. In addition, aspects of attention, such as orienting, sustaining, and shifting, are likely involved in identifying referents to which novel lexical labels can be mapped during word learning.

In this vein, attention, as measured by parent report, has been shown to predict language comprehension, such that increased duration of orienting to objects at 6 and 7 months was related to better receptive language at 10 to 12 months (Dixon & Smith, 2000; Morales et

al., 2000). In addition, experimental tasks have revealed relationships between duration of looking and both later vocabulary ability and word learning in typical development (Dixon & Salley, 2007; Kannass & Oakes, 2008; Salley, Panneton, & Colombo, 2013). For example, Kannass and Oakes (2008) reported a correlation between visual attention in typically developing 9-month-olds and receptive vocabulary at 31 months of age. In toddlers with ASD, visual attention to a novel object has been shown to be associated with later word-learning abilities (McDuffie, Yoder, & Stone, 2006). Taken together, individual differences in aspects of attention have the potential to account for variability in language learning and language outcomes.

The Current Study

Given the evidence of atypical attention in infants with FXS and the link between attention and language development in other populations, visual attention may be a source of variability in the FXS linguistic phenotype, beyond general developmental level. Only one attempt has been made to connect what is known about attention and language in older individuals with FXS, but without empirical data. Addressing language use (i.e., pragmatics), not language structure (i.e., vocabulary, syntax), Cornish, Sudhalter, and Turk (2004) proposed that impaired inhibitory control and hyperarousal lead to the tangential and perseverative language that is characteristic of FXS. There is a pressing need to build profiles of strengths and weaknesses in neurodevelopmental disorders using trajectories of attentional processes (Scerif, 2010). Directly testing the relationship between attention and language in infants with FXS can serve as a first step in this line of research.

We examined the association between direct assessment and parent-report measures of visual attention and later language ability, with an emphasis on vocabulary—the domain of language most likely to be impacted by attention to objects—in infants with FXS. Given the lack of research on infant language and communication in FXS, the goal of the current study was to set the stage for future research by broadly characterizing performance of a small sample of infants with a range of assessments, reporting primarily exploratory analyses in terms of associations among measures, change over time, extent of delay, and the role of visual attention as a predictor.

We addressed the following research questions: **(1) To what degree are direct assessment and parent-report measures of language associated in infants with FXS?** We tested correlations among multiple assessments of emergent language and communication. We expected direct assessment and parent-report measures to be related, particularly within modalities (i.e., receptive language, expressive language). From a methodological perspective, knowing the extent to which various measures of language and communication ability (i.e., parent report and direct assessment) align for young children with FXS would inform the selection of appropriate assessments, both in clinical and research settings. **(2) What is the profile of change in early language ability in infants with FXS?** Early language profiles were characterized in terms of patterns of change in raw scores (i.e., absolute level of ability) and standardized scores (i.e., performance relative to age expectations). We predicted that increased ability, albeit slower than typical acquisition, would be observed in infants with FXS over time between 9 and 24 months of age. **(3) How**

do the language abilities of young males with FXS compare to typically developing males with similar ages or developmental levels? To achieve chronological age- and developmental level-comparisons, we compared the performance of 12-month-old infants with FXS and 18-month-old infants with FXS to 12-month-old typically developing infants. We predicted that infants with FXS would demonstrate delays in language ability relative to both chronological age and developmental level. **(4) Does visual attention predict later language ability in infants with FXS?** We tested visual attention, directly and using parent report, as a predictor of language and communication. We predicted that visual attention would account for individual differences in language and communication ability and, in particular, vocabulary size—even when accounting for developmental level. Our general expectation was that more typical patterns of attention (e.g., shorter looking duration and shorter disengagement latencies during direct assessment) would predict better language skills.

Method

Participants

Data were collected as a part of a longitudinal study conducted at the University of North Carolina at Chapel Hill examining early development and family adaptation to FXS (Mirrett et al., 2004; Roberts et al., 2012). The study included 13 males with FXS assessed four times (i.e., at 9, 12, 18, and 24 months of age) yielding 49 assessments, as well as 11 typically developing males assessed at 12 months of age for a total of 24 participants. Genetic status for infants with FXS was verified by genetic report. Typically developing infants were those with no family history of or current developmental concerns. Participants with FXS lived throughout the United States and were recruited through existing and completed studies, FXS family support groups, and FXS parent listserves. Typically developing infants were recruited through flyers posted in community childcare centers. All participants were Caucasian. Mean income for a subset of participants for whom these data were available ($n = 12$, including 11 with FXS) was \$81,000 (Range: \$32,500 – \$150,000). Parents provided written consent.

Measures

Direct assessments in the current study were administered when infants were 9, 12, and 18 months old. Parent report measures were administered at 9, 12, 18, and 24 months of age.

Attention—Attention was assessed through two complementary measures: direct assessment and parent report. The Laboratory Temperament Assessment Battery (LabTAB; Goldsmith & Rothbart, 1999) is a standardized experimental instrument designed to observe differences in individuals' temperament. We used the pre-locomotor version for infants. Given our interest in attention, we administered the “toy play” experiment in which the child is given a toy key ring to play with for 3 minutes while seated in a high chair or booster seat. Children must be in a neutral state and demonstrate initial interest evidenced by looking at or manipulating the toy as conditions of task completion. Behavioral data from the LabTAB were coded using “The Observer” (Noldus, 1991), a software system for coding and analyzing frequencies and durations of events. The use of this system permits coding

behavior at different playback speeds, while maintaining a time reference for exact coding of behavior initiation and conclusion. To establish reliability, training was conducted until research staff achieved 80% accuracy on at least three consecutive videotapes and an interobserver agreement rate of 80% or higher was maintained throughout the coding process. To maintain reliability, a master coder checked 20% of episodes, maintaining a kappa of .80 or higher. The proportion of total time the child spent gazing at the keys (hereafter, gaze to stimulus) and latency to shift away from the keys were the variables of interest and were taken as indicators of visual attention.

The second measure of attention was the Infant Behavior Questionnaire (IBQ; Rothbart, 1981). The IBQ is a parent-report inventory designed to measure temperament in infants from 3 to 12 months of age. On a 7-point scale, parents are asked to rate their child's behavior within the past week or two. The subscale of interest for the IBQ is Duration of Orienting, which indexes an infant's attention to and/or interaction with a single object for extended periods of time. Reliability estimates for the IBQ range from .67 to .85, with convergent, discriminant, and continuity of temperament dimensions over time supported by Putnam et al. (2008). Because the IBQ was only designed for use up to 12 months of age, we also included an extension of the IBQ, based on the Toddler Behavior Questionnaire (TBQ; Goldsmith, 1996) at the 18- and 24-month assessments. Developed to be compatible with the IBQ, Rothbart and colleagues revised the TBQ by adding 11 additional scales, yielding 16 scales, including the two of primary interest: Attention Focusing and Attention Shifting (Putnam, Ellis & Rothbart, 2001). A final version of this measure is known as the Early Childhood Behavior Questionnaire for children 18 to 36 months of age (ECBQ; Putnam, Garstein, & Rothbart, 2006). On Rothbart's revised TBQ, Attention Focusing refers to the ability to resist distraction for a sustained duration. Attention Shifting refers to the ability to transfer attentional focus from one activity to another. Reliability estimates range from .78 to .89 with evidence for adequate convergent and discriminant validity (Putnam et al., 2008).

Developmental level—The Mullen Scales of Early Learning (MSEL; Mullen, 1995) is a standardized instrument used to assess cognitive and motor abilities in young children from birth to 69 months of age. The MSEL was chosen because of its strong reliability and validity and high correlations ($r = .70$) with the Bayley Scales of Infant Development (Bayley, 1993). The MSEL consists of five subscales: Gross Motor, Fine Motor, Receptive Language, Expressive Language, and Visual Reception domains. The latter three were of interest in the current study, with performance on the Visual Reception subtest taken as an index of general, but principally nonverbal, developmental level. T-scores and age-equivalent scores are calculated based on raw scores for each subtest.

Language and communication scales—Six measures assessed emergent language and communication, which we view as tightly coupled domains at the early stage of development under investigation. Two measures were direct assessments: the MSEL, described above, and the Early Language Milestones Scale-2 (ELM-2). The remaining measures were based on parent report. Descriptive statistics for language and communication performance are presented in Table 1.

The ELM-2 (Coplan, 1993), designed for early detection of speech and language delays, is a screening measure for children from birth to three years. The ELM-2 is a 41-item scale with three domains: auditory receptive, auditory expressive, and visual. Item responses are elicited using history, direct testing, or incidental observation by the examiner, such that items failed by history are directly tested when allowable (11 items), and an item may be passed by incidental observation at any point when allowable (34 items). Items are scored pass/fail based on established emergence ages for each item and then quantified into raw, age-equivalent, and standard scores. The ELM-2 has adequate reliability ($r = .77 - .94$) and validity ($r = .51 - .66$; Coplan & Gleason, 1993).

The Receptive-Expressive Emergent Language Scale, 2nd Edition (REEL-2; Bzoch & League, 1991) assesses language from birth to 36 months, using observational parent report. This scale is comprised of an overall total language development score that yields an age-equivalent and quotient score, and two subscales of receptive and expressive language, yielding receptive and expressive language age-equivalent and quotient scores. This measure has adequate psychometric properties: reliability estimates range from .75 to .80 (Bzoch & League, 1991). Unlike the other parent-report measures, data for the REEL-2 were available only at 9, 12, and 18 months of age.

The MacArthur Communicative Development Inventories: Words and Gestures form (CDI; Fenson et al., 1993) was designed to characterize early language in children ages 8 to 18 months, specifically vocabulary comprehension and usage. Validity (convergent, concurrent, and predictive) and reliability correlations are modest to high (Fenson et al., 1993). These skills are quantified by number of words understood, number of words produced, phrases understood, and gestures used. Number of words understood and produced were of primary interest.

The Vineland Adaptive Behavior Scales, Survey Interview Form (VABS; Sparrow, Balla & Cicchetti, 1984) is a 265-item parental report measure administered through a semi-structured interview for individuals from birth to adulthood. There are four domains on which the VABS provides standardized scores: Communication, Daily Living, Socialization, and Motor. Scores from the VABS were standardized based on a representative sample with a mean score of 100 and a standard deviation of 15. The VABS has adequate test-retest reliability ranging from .76 to .93 and inter-rater reliability ranging from .62 to .78 (Rosenbaum et al., 1995). Of current interest was the Communication domain, including Receptive and Expressive Communication subdomain scores.

The Communication and Symbolic Behavior Scales-Caregiver Questionnaire (CSBS; Wetherby & Prizant, 1990) is a 25-item qualitative measure appropriate for children between 6 and 24 months of age. Predictive validity of the CSBS ranges from .67 to .75 and test-retest reliability is estimated at .86 (Wetherby et al., 2002). Parents are asked to describe their child's communication and symbolic behavior pertaining to daily activities and the ways in which a child communicates his or her needs. Scores include domain composites for social, speech, and symbolic skills and a standardized total score ($M = 100$, $SD = 15$). Total scores were of current interest.

Procedure

Participants were assessed in the family's home for the 9- and 18-month assessments with the 12-month assessment conducted at the research laboratory at the University of North Carolina at Chapel Hill. The 24-month assessment was conducted via telephone interviews and questionnaires. Direct child assessments were done over a span of two days to maximize compliance and to allow for repeated administration attempts as needed. The standard assessment sequence was to complete the MSEL and parent questionnaires on the first day and then to conduct the LabTAB and other direct assessments on the second day. Parent questionnaires were mailed to mothers approximately two weeks prior to, and collected at, the face-to-face assessment.

Results

Given the small sample sizes, nonparametric statistics are reported throughout.

Preliminary Results: Direct Assessment and Parent Report of Attention

LabTAB performance—For infants with FXS, mean proportion of gaze to stimulus was .65 ($SD = 0.28$) at 9 months, .73 ($SD = 0.21$) at 12 months, and .68 ($SD = 0.19$; $n = 11$) at 18 months; mean latency to shift away from stimulus in seconds was 21.26 ($SD = 19.96$; $n = 10$) at 9 months, 32.52 ($SD = 43.93$; $n = 12$) at 12 months, and 19.06 ($SD = 21.65$; $n = 11$) at 18 months. For infants with typical development at 12 months, mean proportion of gaze to stimulus was .54 ($SD = 0.19$) and latency to shift was 9.68 ($SD = 6.07$). At 12 months, similar to findings reported by Roberts et al. (2012), gaze to stimulus was greater in infants with FXS than typical development, $U = 37.00$, $Z = -1.99$, $p = .047$, $r = -.41$, but latency did not differ, $U = 39.00$, $Z = -1.66$, $p = .104$, $r = -.35$.

IBQ scores—For infants with FXS, mean Duration of Orienting was 4.23 ($SD = 1.04$; $n = 10$) at 9 months and 3.73 ($SD = 0.99$; $n = 12$) at 12 months. For infants with typical development, mean Duration of Orienting was 3.61 ($SD = 1.44$; $n = 9$) at 12 months. At 12 months, Duration of Orienting did not differ between infants with FXS and those with typical development, $U = 49.50$, $Z = -0.32$, $p = .754$, $r = -.07$. We also referenced the Duration of Orienting scores to those reported by previous research on typically developing infants: $M = 3.92$ at 3 to 6 months, $M = 3.60$ at 6 to 9 months, and $M = 3.49$ at 9 to 12 months (Garstein & Rothbart, 2003); $M = 3.51$, $SD = 1.08$ at 7 months, $M = 3.57$, $SD = 1.04$ at 10 months, $M = 3.28$, $SD = 0.83$ at 13 months (Dixon & Smith, 2010); $M = 4.05$, $SD = 0.98$ at 6 months (Morales et al., 2000). Descriptively, the mean scores from infants with FXS decrease with age until 12 months, in line with studies on typical development.

TBQ scores—For infants with FXS, mean Attention Focusing was 3.78 ($SD = 0.62$; $n = 10$) at 18 months and 3.96 ($SD = 1.00$; $n = 8$) at 24 months; mean Attention Shifting was 4.45 ($SD = 0.85$; $n = 9$) at 18 months and 4.23 ($SD = 0.74$; $n = 8$) at 24 months. The TBQ was not administered to parents of typically developing children. Thus, as a point of comparison, we referenced these scores to those reported by Putnam et al. (2006) for the ECBQ: Attention Focusing ($M = 4.08$, $SD = 0.85$ at 18 months; $M = 4.38$, $SD = 0.71$ at 24 months) and Attention Shifting ($M = 4.47$, $SD = 0.67$ at 18 months; $M = 4.59$, $SD = 0.68$ at 24 months).

24 months). Descriptively, the observed scores from infants with FXS for Attention Focusing increased from 18 to 24 months, aligning with previous research on typical development, but the pattern for Attention Shifting did not.

In summary, visual attention in 12-month-olds with FXS differed from 12-month-olds with typical development based on direct assessment of proportion of gaze to the stimulus from LabTAB, but did not differ in terms of LabTAB latency to shift or IBQ Duration of Orienting.

Associations among Measures

We examined concurrent nonparametric Spearman correlations among language measures for 12-month-olds with FXS, as shown in Table 2. We chose to interpret only moderate to strong correlations (i.e., greater than .60) given the exploratory nature of our study and the large number of correlations calculated. All significant correlations were positive and a number of measures were moderately correlated, even between direct assessments (i.e., MSEL, ELM-2) and parent reports. The CSBS, reflecting both receptive and expressive skills, was correlated with many scores (i.e., 5 over the .60 threshold). Notably, CDI words understood scores were not correlated with any other language measure. In summary, even at 12 months of age, many direct assessment and parent-report measures of language and communication yield correlated scores for children with FXS.

Change Over Time

We tested change over time using nonparametric Friedman tests for related samples.

Direct assessments—MSEL receptive, $\chi^2(2) = 20.83$, $p < .001$, and MSEL expressive, $\chi^2(2) = 19.62$, $p < .001$, raw scores increased from 9 to 18 months, whereas receptive, $\chi^2(2) = 12.18$, $p = .002$, and expressive, $\chi^2(2) = 8.91$, $p = .012$, T-scores decreased from 9 to 18 months. ELM-2 receptive, $\chi^2(2) = 11.29$, $p = .004$, and ELM-2 expressive, $\chi^2(2) = 8.65$, $p = .013$, raw scores significantly increased over 9 to 18 months. ELM-2 standard scores did not change significantly from 9 to 18 months for receptive, $p = .093$, or expressive language, $p = .326$. Based on direct assessments, receptive and expressive language ability increased for the MSEL and ELM-2, as did extent of delay relative to age expectations for the MSEL.

Parent-report measures—REEL-2 receptive, $\chi^2(2) = 20.46$, $p < .001$, and REEL-2 expressive, $\chi^2(2) = 19.62$, $p < .001$, age-equivalent scores increased from 9 to 18 months, whereas receptive, $\chi^2(2) = 6.19$, $p = .045$, and expressive, $\chi^2(2) = 12.61$, $p = .002$, quotients decreased with time. Infants with FXS demonstrated significant positive change from 9 to 24 months for CDI words understood, $\chi^2(3) = 16.65$, $p = .001$, although the result for failed to reach significance for CDI words produced, $p = .058$. VABS receptive, $\chi^2(3) = 24.97$, $p < .001$, and VABS expressive, $\chi^2(3) = 22.25$, $p < .001$, raw scores increased from 9 to 24 months, whereas the overall VABS Communication standard score decreased, $\chi^2(3) = 24.96$, $p < .001$. CSBS raw scores increased from 9 to 24 months, $\chi^2(3) = 20.74$, $p < .001$, and the decrease in standard scores just failed to reach significance, $p = .051$.

In sum, we detected significant developmental gains in language abilities in the receptive and expressive modalities, as well as significant increases in the extent of delay (i.e., decreasing age-referenced scores), for most language assessments.

Comparisons with Typically Developing Infants

Extent of language delay was examined by comparing MSEL receptive and expressive scores between 12-month-olds with typical development and (1) 12-month-olds with FXS, matched on chronological-age and (2) 18-month-olds with FXS, matched on developmental level. We established equivalence between groups following Kover and Atwood (2013) using Mann-Whitney *U* tests. Participants' performance on the MSEL subtests of interest is shown in Table 3.

Chronological-age comparison—We compared MSEL receptive and expressive raw and standard scores for 12-month-olds with FXS ($n = 13$) and 12-month-olds with typical development ($n = 11$), who did not differ in chronological age, $U = 43.50$, $Z = -1.85$, $p = .106$, $r = -.38$, $s^2_{\text{ratio}} = 0.07$. Participants with typical development had significantly better performance than participants with FXS for receptive language raw scores, $U = 5.00$, $Z = -3.92$, $p < .001$, $r = -.80$, receptive language T-scores, $U = 4.50$, $Z = -3.91$, $p < .001$, $r = -.80$, expressive language raw scores, $U = 1.00$, $Z = -4.12$, $p < .001$, $r = -.84$, and expressive language T-scores, $U = 0.50$, $Z = -4.13$, $p < .001$, $r = -.84$. That is, 12-month-old infants with FXS performed below chronological-age expectations for receptive and expressive language.

Developmental-level comparison—We next compared MSEL language scores for 12-month-olds with typical development ($n = 11$) and 18-month-olds with FXS ($n = 13$), given that their MSEL Visual Reception raw scores did not differ, $U = 48.50$, $Z = -1.35$, $p = .186$, $r = -0.28$, $s^2_{\text{ratio}} = 0.71$. Those with typical development scored higher than those with FXS for receptive language raw scores, $U = 42.50$, $Z = -1.70$, $p = .047$, one-tailed, $r = -.35$, receptive language T-scores, $U = 1.00$, $Z = -4.10$, $p < .001$, $r = -.84$, expressive language raw scores, $U = 29.50$, $Z = -2.51$, $p = .013$, $r = -.51$, and expressive language T-scores, $U = 0.00$, $Z = -4.18$, $p < .001$, $r = -.85$. Thus, language skills of 18-month-olds with FXS were lower than developmental-level expectations.

In summary, receptive and expressive language were delayed in 12-month-olds with FXS relative to chronological age and developmental level. Although the groups were not ideally matched, these comparisons were conservative ones because the mean age and mean Visual Reception raw score of the boys with FXS were descriptively higher than that of boys with typical development and not vice versa. Thus, language weaknesses in participants with FXS relative to typical development are unlikely attributable to younger age or lower nonverbal cognitive skills.

Visual Attention as a Predictor of Language

Table 4 presents longitudinal correlations between visual attention scores, assessed at 12 or 18 months, and language and communication outcomes, assessed at 18 or 24 months for those with FXS. We used nonparametric Spearman correlations and, for significant bivariate

correlations, we followed up with partial nonparametric correlations controlling for developmental level. Again, we focused on the magnitude of correlations with a conservative threshold set at .60 and used two-tailed p -values given the exploratory nature of these analyses.

Direct assessment—Based on LabTAB performance, gaze to the stimulus at 12 months negatively correlated with 24-month CDI scores for words understood and words produced, as well as total scores on the CSBS. Latency to shift at 12 months negatively correlated with 24-month CDI scores for words produced with a correlation of $-.64$, but a two-tailed p -value of .064. Thus, a higher proportion of time gazing at the toy keys, which has been shown to be aberrant in infants with FXS (Roberts et al., 2012), was related to poorer outcomes: fewer words understood and produced, and poorer general communication.

Parent report—Rothbart IBQ Duration of Orienting at 12 months positively correlated with ELM-2 and REEL-2 receptive language raw scores at 18 months and with VABS expressive language scores at 24 months. Rothbart TBQ Attention Focusing at 18 months was positively correlated with VABS expressive language raw scores at 24 months. Overall, higher parental ratings on aspects of attention predicted better language outcomes.

Results from partial Spearman's rank correlations controlling for raw scores on MSEL Visual Reception reflected similar associations among variables, with two exceptions. When controlling for developmental level at 12 months, the relationship between gaze to stimulus and CDI words understood was $-.64$, but had a two-tailed p -value of .066. Gaze to stimulus remained a negative predictor of CDI words produced, $r_{s,ab,c} = -.74$, $p = .024$, and CSBS raw scores, $r_{s,ab,c} = -.76$, $p = .017$. The partial correlation controlling for developmental level at 18 months between Attention Focusing and VABS expressive language also failed to reach significance, $r_{s,ab,c} = .69$, $p = .058$. These follow-up partial correlations should be interpreted cautiously relative to the bivariate correlations because of decreased power due to the loss of a degree of freedom.

In summary, the most consistent findings across analyses were the negative correlations between gaze to the stimulus from LabTAB with CDI words produced and with CSBS raw scores. In addition, Duration of Orienting was positively related to later receptive language scores on the ELM-2 and REEL-2, and expressive language scores on the VABS.

Discussion

This study was the first to examine the association between visual attention and language development in infants with FXS, a population known to have significant difficulties with attentional processes and delays in language and communication ability. Although exploratory in nature due to the small sample size, our results suggest that early attention does predict later language development, with visual attention at 12 months associated with some measures of language and communication during the second year of life. Also, we observed significant delays in language development relative to chronological age- and developmental level-expectations—despite the acquisition of new skills over the period of 9 to 24 months—with individual differences that might be explained, in part, by visual

attention abilities. Finally, we reported the performance of infants with FXS from 9 to 24 months of age across many language and communication assessments and found that scores were generally positively and moderately correlated with each other, providing information likely to be useful to both researchers and clinicians given the previously limited characterization of language and communication ability of infants with FXS.

Visual Attention and Early Language

Certainly, there are many factors that impact language and communication development in infants with FXS, including autism symptoms and maternal responsivity (Roberts et al., 2009; Warren et al., 2010). We suggest that atypical attentional processes, and visual attention in particular, may be a predictor of not only general aspects of development for young children with FXS, but also of the development of language abilities. Cornish, Cole, and colleagues (2012) have suggested that poor attention could negatively impact the intellectual abilities of boys with FXS over time. We extend this notion to language development, a domain of cognition that is critical to success in academics and daily functioning and one that is clearly susceptible to impairment in individuals with FXS across the range of the phenotype.

Based on the experimental LabTAB task, we found that duration of gaze to a toy stimulus at 12 months of age negatively predicted vocabulary words understood and words produced according to parent report on the CDI, as well as CSBS raw scores, at 24 months of age. Thus, more aberrant attention, as defined by increased duration of looking during the LabTAB task, was associated with fewer vocabulary words acquired and less advanced communicative behavior. A less robust finding was that increased latency to shift away from the toy stimulus was negatively correlated with vocabulary words produced on the CDI.

In typical development, the visual attention and orienting systems undergo important changes during the first year. In contrast to 2- and 3-month-old typically developing infants, 4-month-old infants are able to disengage from a stimulus, with visual attention becoming more volitional and obligatory—or “sticky”—attention decreasing (Johnson, Posner, & Rothbart, 2001). Young children with ASD have difficulties with disengagement of attention (Landry & Bryson, 2004) and difficulty with disengagement, especially after the first six months of life, has been linked to later ASD diagnosis for infants at high risk (Elison et al., 2013; Zwaigenbaum et al., 2005). For FXS, the attention difficulties documented in childhood and adolescence are also evident during infancy (Roberts et al., 2012; Scerif et al., 2012) and we have observed that individual differences in visual attention relate to later language ability.

The findings based on parent report also provide evidence that early attention relates to later language skills in infants with FXS. These relationships were present across multiple ages and distributed across several measures. In particular, higher orienting scores at 12 months were associated with better receptive language at 18 months and expressive language at 24 months, even controlling for developmental level. The scores for participants with FXS on the IBQ at 12 months did not differ from scores of typically developing infants in the current sample. It may be that more “typical” attention was associated with higher language skills in this sample of infants with FXS.

Previous studies on typical development have identified similar relationships to those observed in the current study, with attention to objects and longer durations of looking at 6 or 7 months of age associated with better receptive language at 10 or 12 months of age (Dixon & Smith, 2000; Morales et al., 2000). Furthermore, increases in duration of orienting from 7 to 10 months of age may relate to expressive language at 20 months in typical development (Dixon & Smith, 2000). These studies suggest that, within the typical range, individual differences in attention as assessed with the parent-report Rothbart scales may be associated with language, such that increased attention to objects predicts more rapid language development.

In contrast, infants from a high-risk sample with an ASD classification at 24 months had higher scores for Duration of Orienting at 12 months of age relative to those who did not go on to receive an ASD classification (Zwaigenbaum et al., 2005). That is, higher scores for Duration of Orienting may not be developmentally beneficial, at least within the context of ASD. It may also be the case that profiles of change over time in visual attention, rather than static assessments, are the most important predictors of development in other domains (Colombo et al., 2009). While some studies have failed to find systematic changes in scores for Duration of Orienting (Dixon & Smith, 2000), others have reported decreased scores over the first year of life or U-shaped patterns of development (Garstein & Rothbart, 2003; Karrass et al., 2002). Full interpretation of the relationship between language and attention measures for infants with FXS will require a typically developing comparison group assessed longitudinally, particularly because changes in patterns of attention may be more informative than absolute levels of attention for understanding individual differences (Colombo et al., 2009).

Indeed, the direction of effects we observed for language outcomes in FXS differed between direct assessment and parent-report measures of attention. It may be that the Rothbart scales designed to tap attention (i.e., Duration of Orienting, Attention Focusing, and Attention Shifting) assess a different dimension of ability in the FXS population than in typically developing children or, more likely, that they reflect different dimensions of attention than direct assessments. Even within direct assessments, differences in the developmental periods and constructs examined have contributed to disparate patterns of results between paradigms: better outcomes tend to be predicted by shorter looking times in infant habituation, whereas better outcomes tend to be predicted by longer looking times in investigating objects (Colombo et al., 2009; Kannass & Oakes, 2008; Salley et al., 2013). The study by Kannass and Oakes (2008) was designed to investigate such a discrepancy in typically developing children. They found that the context of assessment (e.g., presence of a single object vs. multiple objects) resulted in correlations of opposite directions between visual attention and later language, because of the types of attentional processes that were measured (e.g., processing efficiency for the single object, akin to habituation or LabTAB vs. attentional control for multiple objects, perhaps more akin to IBQ scores; Kannass & Oakes, 2008). These differences highlight the value of employing multiple methods to assess attention. Larger longitudinal studies of direct assessment and parent report of attention in infants with FXS and typically developing infants matched on nonverbal cognitive ability will be necessary to further map the trajectory of visual attention to language development for the FXS phenotype.

Extent of Language Delay and Patterns of Change across Measures

The current study identified significant delays in language ability in infants with FXS by 12 months of age in comparison to both typically developing infants of similar chronological age and, at 18 months, relative to developmental level-expectations. This delay in language ability relative to general developmental level was detected even though the 18-month-old infants with FXS had similar raw scores on the Visual Reception subtest of the MSEL to the 12-month-old typically developing infants. This pattern of delay again suggests that language impairments are evident very early in development in individuals with FXS and that there is a great need for vigilance in their early identification and remediation (Mirrett et al., 2004).

Despite the significant delays relative to the typically developing comparison group on a direct assessment measure of language, we identified significant increases in absolute level of abilities across measures of language and communication, including those based on parent report, with the exception of CDI words produced. In conjunction with this, we also observed significant decreases in standard scores for all but two measures. This suggests that infants with FXS acquire language at a slower rate than would be expected given their chronological ages and that they are likely to fall further behind over time. Similar findings have been demonstrated in older children with FXS (Warren et al., 2010) and for other cognitive domains, such as nonverbal cognitive ability (Kover, Pierpont, Kim, Brown, & Abbeduto, 2013). However, no one has reported this decline in rate of language and communication acquisition during infancy across direct assessment and parent-report measures as we have here. Along with the increase in delays over time, we also observed notable variability in language, especially at later developmental points (e.g., 24 months; see Table 1). These individual differences are a hallmark of the FXS phenotype; teasing apart the factors that lead to such within-syndrome variability will continue to be a crucial goal for future research.

Of the language and communication assessments employed, many were positively and moderately correlated with each other for infants with FXS at 12 months of age, particularly when considering the REEL-2 and the CSBS. Despite the many limitations of parent report for assessing language ability in children (Tomasello & Mervis, 1994), scores from these measures were generally correlated with direct assessment measures, extending what is known about the validity of parent-report measures, such as the CDI in typically developing toddlers, late-talking toddlers, and toddlers with ASD, to young individuals with FXS (Heilmann, Ellis Weismer, Evans, & Hollar, 2005; Nordahl-Hansen, Kaale, & Ulvund, 2014). Concurrent validity is a key finding because evidence of the validity of parent-report measures for infants with FXS could save substantial resources for assessing language and communication in future research, thereby simultaneously allowing the collection of data from a larger number of participants and reducing the burden of direct assessment on very young children who may be taxed by long assessment batteries. Nonetheless, it is of note that CDI words understood were not strongly correlated with other language measures at 12 months of age. Future studies should further evaluate the utility of direct assessment and parent-report measures for assessing specific aspects of language ability in infants and

young children with FXS, with the goal of accurately delineating trajectories of early language acquisition in this population.

Limitations and Future Directions

Related to measurement and analysis, growth scores rather than raw scores or age-equivalents would be preferred as variables used to analyze the developmental trajectories of children with FXS, given the statistical limitations of age-equivalent scores in particular (Kover, McDuffie, Hagerman, & Abbeduto, 2013; Maloney & Larrivee, 2007; Mervis & Klein-Tasman, 2004). In addition, our sample sizes were very small and our statistical analyses were thereby limited. A challenge to future research in this area will be to follow large samples of infants and young children with FXS prospectively to be able to account for the wide variability among individuals in both specific nonverbal cognitive skills (e.g., visual attention) and language ability. While visual attention may be a factor in explaining within-syndrome variability, it will be important to understand the extent to which any observed associations between visual attention and language development reflect syndrome specificity. Cross-syndrome comparisons will be of value to identify the extent to which patterns of development are specific to FXS or are common among neurodevelopmental disorders, including ASD (Abbeduto et al., 2007; Luyster et al., 2011). Differences in visual attention, and perhaps in lower-level volitional eye-movement control, could have implications for not only what we know about language development in populations of children with language impairment, but also how we assess it (Kelly, Walker, & Norbury, 2013).

The overlap between FXS and ASD also provides a challenge. Given their association in infants with FXS, the interplay between visual attention and symptoms of ASD, as they relate to language learning mechanisms, should be examined (Roberts et al., 2012). In the current study, we did not examine joint attention (i.e., triadic orienting between the child, another individual, and an object or event), which is impaired in individuals with ASD and is also related to language ability (Dawson et al., 2004; Toth, Munson, Meltzoff, & Dawson, 2006; but see McDuffie, Yoder, & Stone, 2005). For children with typical development, ASD, or Down syndrome, it may be attention to objects or events that are also the focus of attention of a communicative partner that is most critical for future receptive and expressive vocabulary ability—a topic that has yet to be explored in infants and toddlers with FXS (Adamson et al., 2009).

Because one of the more robust associations with visual attention was found for expressive vocabulary size, word-learning mechanisms will be of particular interest. For young children with ASD, visual attention to objects is associated with associative word learning, with this fast-mapping ability thought to mediate the relationship between attention and vocabulary (McDuffie et al., 2006). Because vocabulary acquisition begins early in life and is foundational to other domains of language, it will be important for future research to discern how aspects of visual attention impact word learning. Eye gaze measures of language processing and learning, as employed for the study of language in children with ASD (Naigles, Kelty, Jaffery, & Fein, 2011; Venker, Eernisse, Saffran, & Ellis Weismer, 2013),

may have utility for assessing children with FXS, particularly for language-learning processes related to lexical acquisition.

Conclusions

Ultimately, the goal of characterizing profiles of specific language domains and their predictors is to identify the mechanisms of learning that contribute to language impairments in children with FXS so that effective remediation will be possible. The purpose of the current study was to provide the foundation for such work. Our exploratory findings require replication; however, strengths of the current study include its longitudinal design and assessment of language and attention using multiple measures. Despite delays in language ability relative to chronological age- and developmental level-expectations, we found that language scores of infants with FXS increased from 9 to 24 months of age with moderate correlations among many measures. The longitudinal associations we identified between visual attention and later language abilities suggest that future research should be aimed at identifying the language learning mechanisms through which visual attention impacts language acquisition in infants with FXS, with a particular emphasis on vocabulary acquisition. Attentional processes could be a critical source of individual differences in the highly variable FXS linguistic phenotype.

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Table 1

Language Performance of Infants with Fragile X Syndrome

Variable	9 months (<i>n</i> = 11)			12 months (<i>n</i> = 13)			18 months (<i>n</i> = 13)			24 months (<i>n</i> = 12)		
	<i>M</i>	(<i>SD</i>)	Range	<i>M</i>	(<i>SD</i>)	Range	<i>M</i>	(<i>SD</i>)	Range	<i>M</i>	(<i>SD</i>)	Range
Chronological age	9.40	(0.54)	8–10	12.21	(0.36)	12–13	17.96	(0.54)	17–19	25.67	(3.45)	24–36
ELM-2												
Receptive raw	5.91	(1.14)	4–8	6.00 ^a	(0.74)	5–7	7.15	(1.07)	5–9	-	-	-
Receptive SS	90.91	(13.72)	69–113	79.83 ^a	(8.54)	69–92	78.60 ^c	(5.97)	69–90	-	-	-
Expressive raw	6.09	(1.14)	5–8	6.33 ^a	(0.99)	4–7	7.46	(1.20)	6–10	-	-	-
Expressive SS	82.18	(13.41)	69–100	77.40 ^c	(5.80)	69–81	75.64 ^b	(9.66)	69–94	-	-	-
REEL-2												
Receptive AE	6.64	(1.43)	5–10	7.38	(1.66)	5–10	10.85	(2.91)	6–16	-	-	-
Receptive Q	69.09	(14.90)	50–100	59.77	(12.24)	42–77	60.31	(16.28)	35–89	-	-	-
Expressive AE	5.91	(0.94)	5–8	6.31	(1.70)	3–10	9.00	(2.61)	6–14	-	-	-
Expressive Q	61.45	(9.21)	50–80	51.08	(13.21)	25–77	49.92	(14.64)	33–78	-	-	-
CDI words												
Understood	17.45	(28.62)	0–97	25.67 ^a	(29.63)	0–96	50.33 ^a	(50.64)	2–149	104.90 ^c	(93.98)	10–299
Produced	0.45	(1.21)	0–4	0.83 ^a	(1.75)	0–6	2.25 ^a	(3.25)	0–10	12.90 ^c	(14.49)	0–34
VABS												
Receptive raw	6.36	(2.11)	4–10	9.54	(2.57)	6–14	13.54	(3.46)	8–20	17.00	(3.05)	12–20
Expressive raw	4.73	(0.91)	4–6	4.92	(1.26)	4–8	6.85	(1.63)	4–9	8.83	(3.33)	5–18
Comm. SS	95.73	(6.86)	88–107	91.15	(6.12)	83–101	76.92	(6.22)	68–90	72.75	(3.62)	67–78
CSBS												
Total raw	31.91	(13.42)	20–62	37.85	(13.92)	17–69	56.67 ^a	(23.15)	14–92	72.40 ^c	(21.59)	36–101
Total SS	89.55	(14.98)	77–122	77.08	(11.84)	66–102	76.64 ^b	(8.48)	68–92	75.40 ^c	(8.20)	65–88

Note. ELM-2 = Early Language Milestones Scale-2; REEL-2 = Receptive-Expressive Emergent Language Scale, 2nd Edition; CDI = MacArthur-Bates Communicative Development Inventories: Words and Gestures; VABS = Vineland Adaptive Behavior Scales; MSEL = Mullen Scales of Early Learning; CSBS = Communication and Symbolic Behavior Scales; SS = standard score; AE = age-equivalent score; Q = quotient score.

^aData were available for only 12 participants.

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Data were available for only 11 participants.
Data were available for only 10 participants.

Spearman Correlations among Raw Scores from Language Assessments for Infants with Fragile X Syndrome at 12 months of Age (n = 13)

Table 2

Variable	1	2	3	4	5	6	7	8	9	10	11	12
1. Age	—											
2. MSEL receptive	.14	—										
3. MSEL expressive	.50 [*]	.71 ^{**}	—									
4. ELM-2 receptive	-.15	.58 [*]	.62 [*]	—								
5. ELM-2 expressive	-.39	.12	-.03	.50 [*]	—							
6. REEL-2 receptive ^a	.03	.79 ^{**}	.76 ^{**}	.78 ^{**}	.50 [*]	—						
7. REEL-2 expressive ^a	.02	.58 [*]	.55 [*]	.64 [*]	.75 ^{**}	.77 ^{**}	—					
8. CDI understood	.17	.29	.17	.14	.01	.35	-.09	—				
9. CDI produced	.32	.56 [*]	.44	.37	.33	.61 [*]	.54 [*]	.31	—			
10. VABS receptive	.04	.05	.31	.50 [*]	.45	.55 [*]	.47	.11	.30	—		
11. VABS expressive	-.20	.55 [*]	.31	.24	.32	.67 ^{**}	.51 [*]	.36	.47	.26	—	
12. CSBS	-.13	.65 ^{**}	.58 [*]	.62 [*]	.16	.78 ^{**}	.61 [*]	.29	.26	.49 [*]	.73 ^{**}	—

^a Age-equivalent scores.^{*} $p < .05$, one-tailed.^{**} $p < .01$, one-tailed.

Table 3
Mullen Scales of Early Learning (MSEL) Scores of Infants with Typical Development or Fragile X Syndrome

Variable	Typical Development			Fragile X Syndrome		
	12 months (<i>n</i> = 11)			12 months (<i>n</i> = 13)		
	<i>M</i>	(<i>SD</i>)	Range	<i>M</i>	(<i>SD</i>)	Range
Chronological age	11.64	(1.03)	11–14	12.21	(0.36)	12–13
Visual reception						
Raw	16.00	(2.90)	13–23	12.54	(1.71)	10–15
AE	13.00	(2.90)	10–20	9.54	(1.71)	7–12
T-score	52.45	(10.79)	38–70	36.77	(6.89)	27–47
Receptive language						
Raw	14.64	(2.84)	12–22	10.92	(1.04)	9–12
AE	13.27	(3.47)	10–22	8.92	(1.04)	7–10
T-score	54.18	(9.94)	40–71	35.54	(5.08)	28–45
Expressive language						
Raw	13.64	(2.73)	11–19	9.00	(1.23)	7–11
AE	13.55	(3.14)	10–20	8.00	(1.23)	6–10
T-score	56.00	(11.24)	42–80	33.46	(5.17)	25–42

Note. AE = Age-equivalent score.

Table 4

Longitudinal Spearman Correlations among Visual Attention Predictors at 12 months and Later Language Assessments for Infants with Fragile X Syndrome (n = 13)

Variable	LabTAB			Rothbart IBQ		Rothbart's Revised TBQ	
	Gaze to stimulus	Latency to shift away	Orienting duration	Attention focusing ^a	Attention shifting ^a		
18-Month Outcomes							
MSEL							
Receptive	.04	.20	.52	–	–		–
Expressive	–.11	.34	.46	–	–		–
ELM-2							
Receptive	–.16	–.06	.67*	–	–		–
Expressive	–.39	–.13	.44	–	–		–
REEL-2							
Receptive	–.08	.02	.62*	–	–		–
Expressive	–.33	.01	.50	–	–		–
24-Month Outcomes							
CDI Words							
Understood	–.70*	–.23	.19	<.01	–.43		–.36
Produced	–.76*	–.64	.25	–.28	–.36		–.17
VABS							
Receptive	–.33	.14	.33	–.08	–.17		.42
Expressive	–.26	–.26	.61*	.73*	.42		–.14
CSBS	–.81*	–.53	.29	.02	–.14		

Note. Raw scores were the variables of interest, with the exception of the REEL-2, for which age-equivalent scores were used. Dashes indicate cells for which longitudinal correlations could not be calculated because assessments were concurrent.

^a Assessed at 18 months.

* $p < .05$, two-tailed.